

TRANSORAL SURGICAL RESECTION OF BILATERAL STYLOID PROCESSES ELONGATION (EAGLE'S SYNDROME)

Abdullah Baharudin¹, Ibrahim Rohaida², Abdullah Khairudin²

Department of Otorhinolaryngology, School of Medical Sciences, Universiti Sains Malaysia, Kota Bharu, Kelantan, Malaysia¹

Department of Otorhinolaryngology, Hospital Sultanah Nur Zahirah, Jalan Sultan Mahmud, Kuala Terengganu, Malaysia²

Corresponding author: Assoc Prof. Dr. Baharudin Abdullah MBBS (Mal), MMED ORL-HNS (USM). Department of Otorhinolaryngology—Head and Neck Surgery, School of Medical Sciences, Universiti Sains Malaysia, Kubang Kerian 16150, Kelantan, Malaysia. Tel: +6097676416. Fax: +6097676424. Email: baharudin@kb.usm.my

Case report

SUMMARY

Eagle's syndrome represents a symptomatic styloid process elongation or calcification of stylohyoid or stylomandibular ligament. The

symptoms include the throat pain radiating to ipsilateral ear or foreign body sensation in the pharynx causing odynophagia and dysphagia. It is commonly unilateral and bilateral cases are rare. We report a case of bilateral elonga-

tion of styloid processes treated surgically by transoral approach.

Keywords: Eagle's syndrome, elongated styloid process, neck pain.

1. INTRODUCTION

Elongation of styloid process is defined by Dr W.W. Eagle as styloid process more than 2.5 cm in length. Incidence of styloid process elongation is around 4-7% and only 4% of the patients with elongated styloid process have symptoms (1, 2). Therefore, the actual incidence of Eagle's syndrome accounted as 0.16%. Peak age of incidence is 30 to 40 years old (2, 3, 4). The diversion of elongated styloid process in relation to its surrounding structures account for a wide range of symptoms of Eagle's syndrome.

2. CASE SUMMARY

A 59 year old Malay lady presented to ORL clinic with history of persistent throat pain for 5 months duration. She also complained of foreign body sensation and odynophagia. She had reflux symptoms and halithosis as well. There was no history of recurrent fever. Clinical examination of her throat showed bilateral enlarged tonsils grade 2. No inflammation and no exudates on the tonsils were noted. Howev-

er there was a pale looking mass over superior pole of the left tonsil measured about one by one cm. The same looking mass also present on the right tonsil but smaller. The masses were hard on palpation and tender. She was diagnosed as having bilateral tonsilolith and underwent bilateral tonsillectomy. Intraoperatively, after both of the tonsils were removed, there was a bony hard bulge noted over the left tonsillar fossa by the operative surgeon. She was reviewed 2 weeks post operatively and had some improvement in her symptoms. Histopathology of the removed tonsils reported as chronic inflammation of the tonsils tissue, but no tonsilolith was found. After 3 months she still complaint of persistent throat pain which was even more severe, sharp in nature and exacerbated by swallowing, chewing and jaw movement. However, she did not have any more halithosis and foreign body sensation. Review of her oropharynx showed both of her tonsillar fossa have healed well and there was a prominent bulge on the left tonsillar fos-

sa. Digital palpation over the tonsillar fossa revealed that the bulge was bony hard and exacerbated the left neck pain. No other neck mass was noted. A 3D CT of styloid process was done and showed bilateral styloid processes elongation (Figure 1). She then underwent bilateral styloid processes shortening via transoral approach in which 2.5 cm long of left styloid and 1.5 cm long of right styloid were resected (Figure 2). She was reviewed 1 month later and her symptoms resolved.

3. DISCUSSION

Styloid process arises from temporal bone just anteromedial to stylomastoid foramen. Stylohyoid complex consist of styloid process, stylohyoid ligament and lesser horn of hyoid bone derived from 2nd branchial arch of Reichert cartilage. Three muscles i.e styloglossus, stylohyoid, stylopharyngeus and two ligaments i.e stylohyoid and stylomandibular ligament attached to the styloid process. Styloid process has closed relation to important neurovascular structure in the neck i.e v,

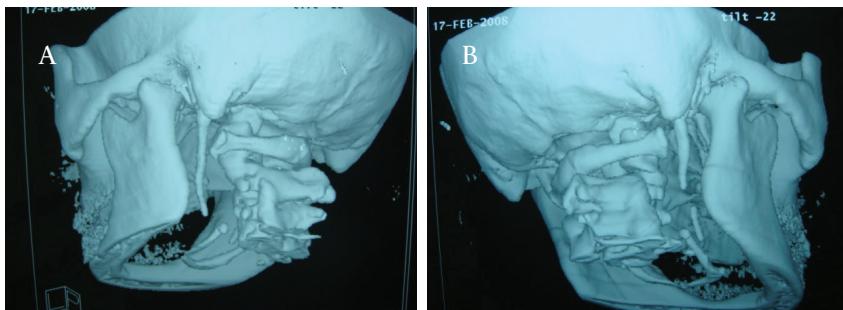


Figure 1. Image of 3D reconstruction of styloid processes showing both styloid processes of the patient to be elongated and pointing medially, 1(a) left styloid process [5cm long] and 1(b) Right styloid process [3 cm long].

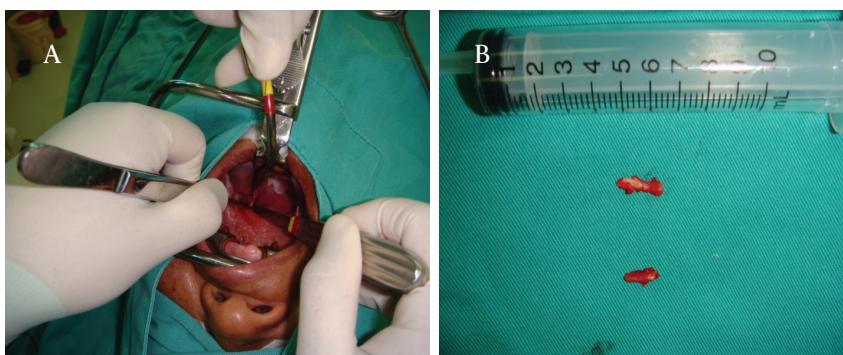


Figure 2. Shortening of both styloid processes of the patient performed via transoral approach , 2(a) skeletonizing of left styloid process, 2(b) part of left styloid process (upper bony specimen) and right styloid process (lower bony specimen) that had been resected

vii, ix, x, xi, xii cranial nerves and internal jugular vein. The tip of styloid usually lies between internal and external carotid arteries.

The variation of diversion of elongated styloid process may be the cause for wide variety of symptoms in Eagle syndrome's patients (2, 3, 5). It can be either medial or lateral angulation and anterior or posterior angulation of styloid diversion. Haluk et al. concluded the length and anterior angulation of the styloid process are responsible for the symptoms rather than medial angulation (6). The most common presenting symptoms are persistent throat pain, foreign body sensation, dysphagia, odynophagia, chronic neck pain, headache and referred otalgia (2, 3, 4, 5, 6, 7). The exact mechanism leading to pain in Eagle's syndrome is still uncleared but several postulations have been given. These include compression of glossopharyngeal nerve, degenerative and inflammatory insertion tendinitis, irritation of pharyngeal mucosa and v,vii, ix and x cranial nerves by fibrosis of post tonsillectomy scar, as well as sympathetic

nerve irritation due to impingement on carotid vessels (1, 5, 7).

The etiology of elongation of styloid process also remains unknown (8). Excessive ossification of the stylohyoid complex may be the cause of lengthening of the styloid process (6, 8, 9, 10). Other theories that have been proposed include congenital elongation of styloid process and growth of osseous tissue at the insertion of stylohyoid ligament (9, 10).

History and physical examinations are crucial in the diagnosis of Eagle's Syndrome. However as the presentations actually can be misleading, the aid of imaging is very useful to confirm the diagnosis. Digital palpation over tonsillar fossa is important as a hard bony prominence can be felt indicates elongated styloid process provided that tonsils tissue are not enlarged or absent as in post-tonsillectomy patient and gagging sensation can be tolerated well (3, 5, 11). As in our reported case, since the patient had enlarged tonsils, the elongated styloid process had pushed the tonsils tissue over the superior pole giving the

appearance of tonsilolith like mass. In fact, it was not a true tonsilolith. This additional finding proved the heterogeneity of presentation in Eagle's Syndrome (4). When there is pale whitish mass appeared over the tonsil, clinicians should bear in mind the possibility of an underlying elongated styloid process.

Elongated styloid process can be visualized on anteroposterior modified Towne's view X-ray (3, 4, 5, 6, 7). Any medial and lateral deviation also can be evaluated. Lateral skull X-ray and orthopantomogram also can visualize the elongated styloid process (2, 5, 10, 11, 12). In any centre where 3D CT is available, it is very useful to subject patients for it as this can provide accurate measurement in the length and deviation of styloid process or any other related anatomical variants before surgical intervention take place (6, 10, 11, 12). Angiography may be also useful in any case suspected for carotid artery impingement in Eagle's Syndrome but seldomly done as it is an invasive procedure (4).

It is generally accepted that surgical resection of elongated styloid process is the primary treatment for Eagle's Syndrome (5, 7, 8, 9, 10, 11, 12, 13). Even though medical treatment using analgesic or local steroid injection is an option, the condition is not yet proven to be well treated or resolved (2). There are 2 surgical approaches that has been implemented i.e transoral and external transcervical approach (7, 8, 9, 10, 11, 12, 13). Transoral approach is well described by W.W Eagle where as external approach by Loeser and Cardwell (5). We choose to do transoral approach for our patient since it is technically easier as patient already underwent tonsillectomy and provided no external scar. The procedure was performed under general anaesthesia. Boyle's Davis mouth gag was inserted for better view of surgical field. The protruberance of elongated styloid process identified by digital palpation and then, the overlying mucosa was incised. Dissection over superior constrictor muscle performed to expose the tip of styloid process and skeletonization done until its origin. The liga-

ments and muscle tendons attached to it were separated and the free styloid process resected using bone nibbler as near as possible to its base. The muscles and mucosa over the surgical bed were closed in layers. Surgical failure rate is around 20% by means of partial relief of symptoms or recurrence of symptoms and can be due to entrapment within fibrous tissue of adjoining nerve, or inadequate shortening leading to constant irritation (5).

4. CONCLUSIONS

Eagle's syndrome is a rare condition. Patient may present with vague symptoms such as chronic neck pain, odynophagia, dysphagia, foreign body sensation or headache. Clinicians should have a high suspicion index and in addition, any bony hard mass over tonsillar region elicited by digital palpation should be investigated for a possible elongated styloid process. Styloidectomy is the treatment for the relieve of symptoms in this condition.

Conflict of interest: none declared.

REFERENCES

1. Chourdia V. Elongated Styloid process (Eagle's Syndrome) & Severe Headache. Ind J of Otolaryngology and Head Neck Surg. 2002; 54(3): 238-241.
2. Kishore Chandra Prasad, M. Panduranga Kamath, K. Jagan Mohan Reddy, Krishnam Raju, Saurabh Agarwal. Elongated Styloid Process (Eagle's Syndrome): A Clinical study. J Oral Maxillofac Surg. 2002; 60: 177-175.
3. Beder E, Ozgursoy OB, Ozgursoy SK. Current Diagnosis and Transoral Surgical Treatment of Eagle's Syndrome. J Oral Maxillofac Surg. 2005; 63: 1742-1745.
4. Abie H. Mendelsohn, Gerald S. Berke, Dinesh K. Cchetri. Heterogeneity in the Clinical Presentation of Eagle's Syndrome. Am Otolaryngology - Head and Neck Surgery. 2006; 134: 389-393.
5. Lal M. Gosh, Siba P. Dubey. The syndrome of elongated styloid process. Auris Nasus Larynx. 1999; 26: 169-175.
6. Yavuz H, Caylakli F, Yildirim T, Ozluoglu LN. Angulation of the styloid process in Eagle's Syndrome. Eur Arch Otorhinolaryngology. 2008; 265: 1393-1396.
7. Diamond LH, Cottrell DA, Hunter MJ, Papageorge M. Eagle's Syndrome: A Report of 4 Patients Treated Using a Modified Extra-oral Approach. J Oral Maxillofac Surg. 2001; 59: 1420-1426.
8. Arkuszewski P, Przygonski A, Tyn-dorf M. Eagle's syndrome- report of rare case of bilateral elongation of styloid processes. Otolarnygol Pol. 2009; 63(2): 162-164.
9. Kiran V. Padayappanavar, PS Bhushreddy, AS. Nagalekar. Case Report on Elongated Styloid process. Al Ameen J Med Sci. 2010; 3(1): 99-101.
10. Ryan D. Murtagh, Jamie T. Caracciolo and Gaspar Fernandez. CT findings associated with Eagle's Syndrome. Am J Neuroradiol. 2001; 22(7): 1401-1402.
11. Subramaniam S, Majid MD. Eagle's syndrome. Med J Malaysia. 2003; 58(1): 139-141.
12. Alcalde RE, Ueyama Y, Nishiyama A, Mizuguchi T, Matsumura T, Kishi K. Diagnostic Imaging of Eagle's Syndrome: Report of Three cases. Oral Radiol. 1994; 10(2): 143-148.
13. Buono U, Mangone GM, Michelotti A, Longo F, Califano L. Surgical approach to the Stylohyoid Process in Eagle's Syndrome. J Oral Maxillofac Surg. 2005; 63: 714-716.